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NEUROLOGICAL PICTURE

Harlequin syndrome



A 37 year old woman presented with a 5 month history of left hemifacial flushing induced by exercise leading to the nickname at her jogging club of half-face. She had also noticed increased warmth of the contralateral arm and sometimes the leg. Sometimes the ipsilateral eye would partially close. There was a history of migraine and ulcerative colitis. Full examination was normal apart from a subtle left ptosis. The photograph was taken after a 25 minute run.

Harlequin syndrome was first coined by Lance and Drummond in 1988 when they described five cases of unilateral flushing and sweating.1 Four of these were induced by exercise and they suggested that the lesion was due to torsional occlusion of the anterior radicular artery at the third thoracic segment. In 1993 they investigated two further patients plus two from the original report. Investigations suggested that the lesion involved both preganglionic or postganglionic cervical sympathetic fibres and parasympathetic neurons of the ciliary ganglion.2

Caparrous-Lefebvre et al described a 45 year old woman with unilateral facial loss of facial flushing and sweating with contralateral anhidrosis induced by exercise.3 She also had tonic pupils and areflexia. This was thought to be due to Ross's syndrome (Adie's syndrome with anhidrosis).

Harlequin syndrome is related to the Holmes-Adie and Ross syndromes and the persisting autonomic neuropathy of Guillain-Barré syndrome.

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